CASE REPORT

Dermoid Cyst of Submental Region Mimicking Pilomatricoma

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Abstract Diagnosis of midline neck swellings may give a myriad of options, ranging from ranula, thyroglossal duct cyst, cystic hygroma, cystic lymphangioma, epidermoid, dermoid and teratoid cysts to benign or malignant neoplasms of the mucosa or salivary glands and also rare skin appendage tumours like pilomatricoma or pilomatrix carcinoma. Various pitfalls are encountered in diagnosing these midline swellings as most of the features are overlapping. Careful history and examination have to be complemented with other diagnostic aids such as conventional radiography, ultrasonography, aspiration cytology and computed tomography to arrive at a correct diagnosis. Dermoid cysts in the head and neck are relatively uncommon, present in the midline either above or below the mylohyoid muscle thus differentiating it into sublingual and submental dermoids and can grow to a massive size before presenting symptoms if it breaches the mylohyoid muscle. Pilomatricoma is a skin appendage tumour which presents as midline swelling and is usually diagnosed with careful history and certain pathognomonic features on fine needle aspiration cytology and ultrasonography. Hereby we are presenting a case of a submental swelling which was diagnosed as a dermoid cyst upon excision but which initially posed a diagnostic challenge because of features of pilomatricoma seen on FNAC.

Keywords Dermoid cyst · Pilomatricoma · Submental swelling · Midline neck swelling

Introduction

Dermoid cysts of the head and neck are uncommon, but have been documented since 1778 [1] and constitute only 6.9 % of all dermoids of which 26 % are in the floor of the mouth [2]. They are believed to arise from failure of the overlying surface ectoderm to separate from underlying structures. Typically, Dermoid cysts of head and neck are known to be slow growing, non tender, situated in the midline submentally or sublingually, present at birth, but most often they are diagnosed in second and third decade of life and they may attain significant size before presenting symptoms. It is distinguished from epidermoid cysts by the presence of an epidermoid lining containing adnexal structures such as sweat glands, sebaceous glands, hairs and hair follicles [3–5]. Pilomatricoma is a tumour derived from the primitive basal cells of the epidermis that differentiate into hair matrix cells and it is described as a benign skin appendage tumour [6].

Case Report

A 39 year old male reported with swelling in the submental region. The swelling which was present since birth progressively attained a size of 6 × 6 cm and had become obvious over the last 6 months. The swelling had not shown any sudden increase or decrease in size. The skin over the swelling appeared normal with no signs of inflammation or ulcerations. The swelling was freely movable, did not show any fixation to underlying or

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Fig. 1 Sagittal sections on CT

overlying structures, was non tender and doughy in consistency on palpation. The swelling was not moving on deglutition. Intra orally there was no relevant findings indicative of any extensions into the floor of the mouth. Tongue movements were normal.

Ultrasonography revealed a well defined thin walled homogenous hypoechoic lesion measuring $59 \times 41 \times 59$ mm in the submental region. Vascular lesions were excluded on Colour Doppler study. There was no evidence of any communication with the thyroid gland and features were suggestive of a benign cystic lesion.

Computed Tomogram of the mandible and the submental region revealed a thin walled hypodense lesion of 10–15 Hounsfields Unit (HU) in the submental region (Fig. 1). The lesion was attached to the hyoid bone posteriorly. The lesion did not show enhancement on post contrast study. There was no evidence of calcification or suppuration or bony changes. All these features were suggestive of a benign cyst in the submental region, possibly a thyroglossal cyst.

Aspiration cytology revealed 0.4 ml thick chalky aspirate, which when subjected to microscopic examination showed areas of ghost like cells with extensive calcifications surrounded by inflammatory cells consisting of neutrophils and eosinophils, some occasional anucleate squames were also seen. These features were suggesting a diagnosis of 'Pilomatricoma'.

Correlating the above findings, a preoperative diagnosis of Pilomatricoma was established and the lesion was excised. The lesion was situated between the geniohyoid and mylohyoid muscle and attached to the hyoid bone below (Fig. 2). A white thick caseous fluid with strands of hair like structure were seen in the cystic cavity (Fig. 3). Surgical wound was closed in layers after placement of drains. The surgical wound healed uneventfully.

Fig. 2 Intra operative view (lining)

Fig. 3 Aspirate and excised specimen

Post operative histopathological examination of the excised specimen showed an ulcerated cyst, lined by stratified squamous epithelium, with the supporting stroma showing sebaceous glands and diffuse mononuclear infiltrate with few congested blood vessels favouring a diagnosis of Dermoid Cyst.

Discussion

The cysts in the midline are hypothesized to be caused due to one of the following:

1. Entrapment of ectodermal tissues of first (mandibular) and second (Hyoid) branchial arches, which is called dysodontogenic hypothesis [7, 8].
2. Traumatic implantation of epithelial cells into deeper tissues which suggests traumatic cause [9].
3. Third theory considers dermoid cyst as an variant of thyroglossal cyst, which was one of the preoperative diagnosis in our case based on findings of C.T. scan.

The cysts of the midline can be classified according to its relationship to the muscles of the floor of the mouth. Sublingual cysts; cysts presenting as swelling in the floor of the mouth and situated above the mylohyoid and genioglossus. Submental cysts present as a swelling below the chin and is situated between the mylohyoid and geniohyoid muscle as was seen in our case. The third variant would be a cyst occupying both sublingual and submental spaces, perforating the oral diaphragm which would be a large cyst [8].

Cases have been reported where there have been near total obstruction of the oral cavity due to giant submental dermoid cyst [10].

Histologically, three different types of midline cysts are seen; epidermoid (simple), dermoid (compound) and teratoid (complex) [7]. Epidermoid cysts are always lined by stratified squamous epithelium without dermal appendages within the underlying connective tissue, whereas dermoid cyst, in addition contains dermal appendages like hair follicles, hairs, sebaceous and sweat glands [11] as was seen in our case. The teratoid or complex cysts exhibit lining and wall with tissue structures derived from all three germ layers; ectoderm, endoderm and mesoderm.

Pilomatricomas are asymptomatic, firm, non tender masses which are usually seen in the subcutaneously plane, sometimes adherent to the skin. Stretching of the skin over the tumour shows the ‘Tent sign’, a pathognomonic feature of pilomatricoma and pressing on one edge of the lesion causes the opposite edge to protrude from the skin in a see-saw effect, known as ‘Teeter-Totter sign’ [12]. Pilomatricoma are erroneously diagnosed as epidermoid cysts in

9.1 % cases [13]. Imaging methods such as ultrasonography and computed tomography has limited utility in diagnosis of pilomatricoma and in our case there were no relevant findings related to pilomatricoma such as calcified nodule or a hypoechoic nodule with internal calcified foci [14]. Presence of ghost cells in FNAC would be highly suggestive of pilomatricoma [15] and in our case we could see ghost cells with extensive calcification in FNAC and these findings mimicked pilomatricoma in our case. Pilomatricomas constitute a benign skin appendage tumour derived from primitive basal cells of epidermis, that differentiate as hair matrix cells [13]. Dermoid cysts also contain adnexal structures such as sweat glands, sebaceous gland, hair and hair follicles [4, 5]. The treatment is complete surgical excision in pilomatricoma and dermoid cyst and recurrence is unlikely after complete excision. In case of Pilomatricoma where skin is adherent to the tumour, the excision is carried out along with the overlying skin [16]. The diagnosis of pilomatricoma should be arrived with great caution, as it can mimic various other benign and malignant conditions such as epidermal inclusion cyst, branchial cleft remnants, lipoma, degenerating fibroxanthoma, osteoma cutis, ossifying hematoma and dermoid cyst as in our case [15, 17, 18].

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